



Atypical Localizations of Hydatid Disease: US and CT Evaluation

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Abstract: Hydatid disease (HD) is a unique parasitic disease which is endemic in Macedonia, most frequent localized in liver and lungs. It appears in the form of solitary or multiple cysts, different in size, homogenous, clearly defined, sometimes accompanied by complications.

In this research, we present our experience with atypical localizations of HD: kidney, brain, muscle-skeletal system, spleen, peritoneum, rectum, uterus, ovary, and breast.

The patients were examined by using conventional radiologic and other imaging methods such as ultrasound (US) and computed tomography (CT).

Renal echinococcosis was found in 7 patients, a brain in 3, skeletal in 2, spleen in 3, gallbladder in 2, peritoneal in 3, muscular in 1, ovarian in 2 and breast in one patient.

Native radiograms showed multilayer calcifications and a soft tissue shadow, while contrast radiograms of the abdomen showed signs of intraperitoneal, extraluminal expansion, pressing the surrounding structures. Intravenous urography (IVU) gives pictures of a repressed channel system by a tumor formation, with signs of obstruction and hydronephrosis.

US imaging has a great value in HD screening, being also cheap easily available, simple and with great diagnostic accuracy. CT imaging gives precise topographic and morphologic characteristics of the pathological process, highly sensitive, specific and also accurate.

We can conclude that imaging methods have a great diagnostic value in HD with atypical localization.

Keywords: echinococcosis, hydatid disease, hydatid cyst, atypical localisations, US and CT.

1. INTRODUCTION

Hydatid disease (HD) is an endemic disease in the Republic of Macedonia, occurs after contact of the person with an infected dog who is a primary host of hydatid disease or after the use of food contaminated with canine feces, which contains eggs from the echinococcus. (1, 2, 3) The infection can be asymptomatic or severe, causing extensive organ damage.

After ingestion, the eggs are released from their casing and the embryo penetrates the mucosa from the duodenum and through the v. Portae comes to the liver, where the embryos are stopped and slowly transforms into a larva of the vesicular type - hydatid cyst (HC). (1,2) The most commonly affected organ is liver (70%), followed by lung (20%) and other organs (10%) such as the peritoneal cavity, spleen, kidneys, brain, bones, genitals, muscles, etc. (4,5,6,7,8,9) Hydatid cyst disease, also known as Echinococcosis, is a zoonotic infection caused by the genus Echinococcus. (1, 2, 3) Humans are an accidental intermediate host.

The outer cyst shell consists of concentric lamellar membranes - a hydatid membrane, and

under it is an inner germinal layer of cells. HC is fulfilled with fluid and numerous small cysts that are created from the germinal layer by the swelling and released into the interior of the HC. Their number can reach up to several hundred thousand. In some organs, HC can be surrounded by fibrous tissue - pericitis or adventitia. (1, 2, 3)

Echinococcus granulosus forms a large HC, which, by dissemination, produces secondary cysts and Echinococcus multilocularis seu alveolaris, which creates a spongy mass that constantly grows, there are many cysts separated by fibrous tissue in its interior. (1, 2, 3)

HC growth is usually asymptomatic and quite slow, the clinical manifestations come from the compression of the affected organs. The process can mimic benign or malignant tumor, single or multiple metastases, abscess, cysts, empyema, and other lesions. (4)

Their recognition is not so simple that clinical and radiological investigations play an important role in the diagnosis of this parasitic disease. (4, 10, 11, 12)

We present our experience with atypical sites of hydatidosis and diagnostic evaluation.

2. MATERIAL AND METHODS

Out of the total number of patients 24, men were 14, and women were 10, aged 5-63 years.

Conventional radiological methods, as well as imaging methods, have been used in all patients. Native radiograms of the lung, skeleton, abdomen, urinary tract and intravenous urography (IVU), barium meal (BM) and barium enema (BE) are made.

IVU was performed in 7 patients, after standard preparation of patients with i.v. application of 30-60 ml. 60% iodine contrast agent, making more radiographs at different time intervals.

Small bowel follow-through (SBFT) and BE were applied in 3 patients, after standard preparation, with the use of standard techniques and procedures, several radiographs were made at different time intervals and in several positions and directions.

Ultrasonography examination (US) of the abdomen, retroperitoneum, and pelvis was performed on Toshiba device, using a 3.75 MHz convex probe, and soft tissues US examination was made using a linear probe of 7.5 MHz. US examination was performed in 18 patients.

Computed tomography (CT) examination of the brain, abdomen, retro peritoneum, pelvis, and limbs was performed on the Siemens somatom DRH device. CT examination was performed in 23 patients. CT was performed in two series: a native and a series after i.v. application of non-ionic contrast medium (Omnipaque) 60 ml. The distance between the scans and the width of the scans is 10 mm, and the distance between the scans and the width of the scan for CT scan of the skeleton is 3 mm.

3. RESULTS

Preoperative diagnosis was established by the clinical examination, serological tests (indirect hemagglutination test), X-ray examination, US and CT examination. Nonspecific abdominal pain and a nontender palpable abdominal mass were the most predominant symptoms, other symptoms varied according to the localization of the HC. Patients with brain HD presented with neurological symptoms. The diagnosis of HC was confirmed on histopathological examination of the specimen in all cases.

In our study, primary renal HD occurs in 7 patients. It can be asymptomatic, wherein most patients the clinical presentation includes lateral

mass, renal colic, persistent fever, haematuria, dysuria, hydaturia, pyuria, renal calculi or hypertension, symptoms that are not specific to diagnosis.

The initial radiogram shows an increased contour of the affected kidney with a soft-tissue mass with scaly calcification. In the IVU dominates the suppression and obstruction of the duct system with subsequent hydronephrosis and parenchymal reduction, and the renal contour is deformed.

The US examination shows cystic tumor mass that deforms the kidney, suppresses the duct system, with subsequent hydronephrosis and parenchyma reduction. Nonfunctional kidney we have in 3 cases. (Figure 1) The HC is clearly limited, and in the interior, there are septas, daughters cyst, and peeled hydatid membranes.

The CT finding consists of a unilocular or multilocular cyst, with a density of 8 to 27 HE, which most often involves both ends, with well-defined walls, which are with contrast enhancement. (Figure 2) Calcifications of the walls and daughters cystic which are with less density are often encountered in the cyst. (Figure 3) The presence of these two findings helps in establishing the right diagnosis.

Primary involvement of the spleen from HD is very rare, we met in 3 cases. Secondary, it is involved in rupture of the HC of the liver or abdominal wall, with dissemination to the spleen. The clinical manifestation is nonspecific and consists of abdominal pain and an enlarged spleen.

US and CT findings consist of cystic spleen changes, fulfilled with fluid, hydatid sand, and cystic debris. (Figure 4) Wall calcifications, as well as multiple cysts, are often encountered in the large cyst.

Cerebral HD is also a rare and non-specific localization of the disease and is even more common in children than in adults in our material we encounter in 3 cases.

The clinical manifestation includes a headache, vomiting, edema of papillae or blurred vision. It is more often supratentorial, encompassing the area of a. cerebri media. The parietal lobe is the most common site of the development of the cerebral HC.

The CT image is typical and consists of a large, well-rounded or oval intraparenchymal lesion, which contains a fluid with a density

approximate to the cerebrospinal fluid. (Figure 6) Perifocal edema is not noticeable despite the severe mass effect and the existing hydrocephalus.

In our study, the primary HD of the musculoskeletal system occurs in 3 patients, 2 on the skeleton and 1 in the soft tissues (muscles). The involvement of the skeleton of primary HD is rare and occurs in well-vascularized bone zones. Most commonly affected are vertebrae, epiphyses of the long bones, iliac bones, skull, ribs and soft tissues. In our material, in 1 case the HC is on the spine, and in 1 case in the long bones. The HC occupies only part of the vertebrae, and the remaining part of the cyst extends into the environment. After i.v. application of a contrast medium we have poor contrast enhancement of the pericyst. The lesions in the long bones are revealed as a consequence of a pathological fracture, and in the spine, the first sign is the compression of the cauda equina and spinal nerves.

CT image of bone lesions is similar to that seen on radiographs, a well-defined, multilocular, osteolytic lesion, with a honeycomb appearance, accompanied by an inflammation of the bone and a thinning of the cortex. CT accurately determines the boundary of destruction and clearly reveals spreading the process outside the bone in the surrounding soft tissues. There may be a typical picture of a cyst, round or oval, with sharp and thin edges, which poorly contrast enhancement, and the cyst content shows a liquid density of 8 - 32 HU. There is a clear border between the HC and the bone, and the bone in the surroundings of the cyst is of normal structure and morphology.

The HC in soft tissues manifests itself with a palpable mass that compresses the affected organs. US findings are used in the diagnosis of soft tissue HD, with cysts being shown in the shape of a grape or soap bubbles of different

sizes. CT findings consist of unilocular or multilocular cysts, with an atypical solid component due to inflammatory changes, so that it can mimic the tumor. (Figure 6)

In a patient at age of 27, we found a primary multilocular HC in the left ovary. CT scan clearly demonstrates well-defined, multilocular HC in shape of soap bubbles of different sizes in the left ovary. (Figure 7) In another patient of age at the age of 19, we found an HC of ovarian, uterus and rectum, which on the US and CT had typical features for the HC. The patient was operated on and after three years of operation, a relapse of the HC on the right ovary was registered with US and CT. (Figure 8)

We have two cases with HC in the gallbladder, one with primary hydatidosis, and the second with a rupture of HC from the hepatic in the biliary ducts and gallbladder. (Figure 9) On US and CT there are two visible HC in the liver. The first one in the right lobe is inflamed with good delineation from the hepatic parenchyma with fibrous tissue, pericystic membrane or adventitia. (Figure 10) The same HC is penetrated into the biliary ducts with maintained fistula and communication with the biliary ducts as well as with gallbladder. The gallbladder is calculous with thickened and septated wall with present gallstones and filled by small hydatid daughter cysts. (Figure 11) The biliary ducts are widened especially the common hepatic duct in which the daughter cysts are seen as a cause for obstructive jaundice.

Peritoneal HD is also rare, in our material it appears in 3 patients, it can be located anywhere and is usually a consequence of the rupture of the hepatic, splenic or mesenteric HC. Cysts can cause abdominal distension or obstruction. On the SBFT and BE examinations of the digestive tube, it shows lateral pushing by the soft-tissue mass. Using US and CT, a clear cystic formation is obtained, clearly bounded by a thin wall, septated, with daughter cysts present. (Figure 12)

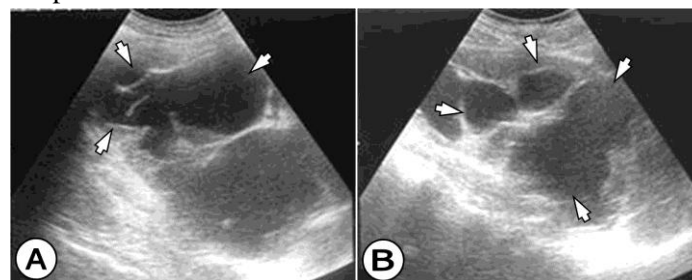


Figure1. (A and B) The US of the kidney: Multilocular HC of the kidney (arrows) seen with obstruction of the channel system and parenchymal reduction.

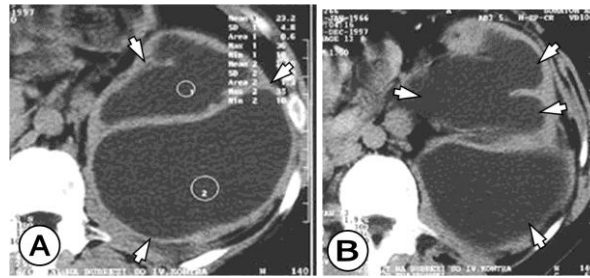


Figure2. (A and B) Contrast-enhanced CT scan of the kidney axial view: We can see, a non-functional kidney with a multilocular HC (arrows). There is an almost complete reduction of the kidney parenchyma with hydronephrosis.



Figure3. Contrast-enhanced CT scan of the kidney axial view: We can see a non-functional kidney with a multilocular HC (arrows) in which clearly visualized detached germinal membrane (arrowhead). There is an almost complete reduction of the parenchyma with hydronephrosis.

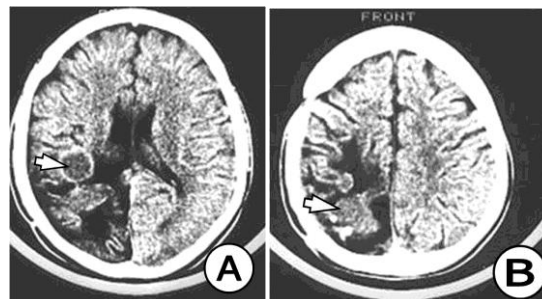


Figure4. (A and B) Unenhanced CT scan of the brain axial view: CT scan demonstrates well-defined, smooth, thin-walled round cyst (arrow) in the right parietooccipital region without causing mass effect and midline shift to the left. There is a recurrent HC of the brain.

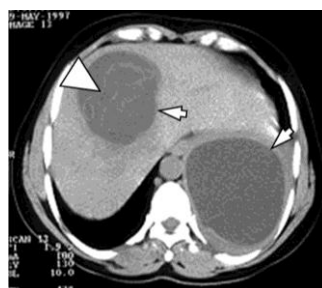


Figure5. Unenhanced CT of abdomen axial view: CT scan demonstrates well-defined, smooth, thin-walled round solitary HC of the spleen (arrow). In the right lobe of the liver, HC is clearly visualized with the detached germinal membrane (arrowhead).

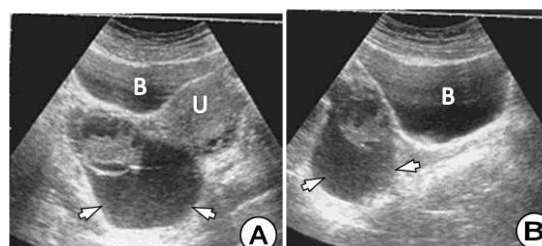


Figure6. (A and B) the US of the pelvis: There is a recurrent HC to the right ovary (arrows) two years after surgery. B-bladder and U-uterus.



Figure7. Contrast-enhanced CT scan of the of abdomen and pelvis axial view: CT scan clearly demonstrates well-defined, multilocular primary HC in the left ovary (arrows).



Figure8. Contrast-enhanced CT scan of the abdomen and pelvis axial view: CT scan clearly demonstrates well-defined, round, solitary primary peritoneal HC in the left part of the pelvis (arrows).

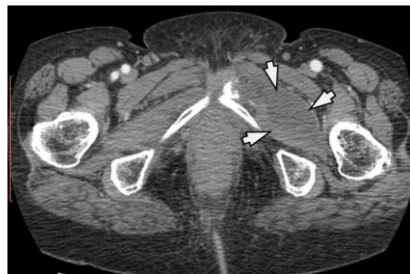


Figure9. Contrast-enhanced CT scan of the pelvis - axial view: CT scan clearly demonstrates well-defined, solitary primary HC in the left part of the pelvis with the destruction of the left pubic bone (arrows).

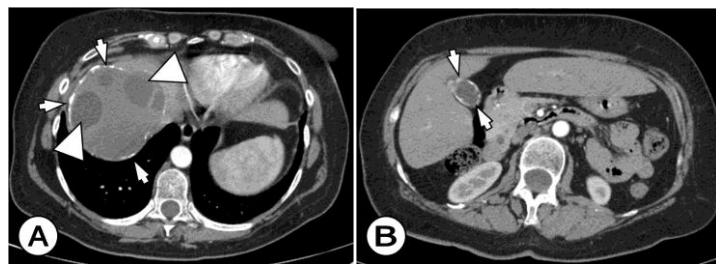


Figure10. (A and B) Contrast-enhanced CT scan of the upper abdomen - axial view: A) in the right lobe of the liver CT scan shows, a giant unenhanced HC with well-defined borders and calcified walls (arrows). Multiple round daughter cysts are seen peripherally within the lesion (arrowhead). B) Inthe gallbladder, we can see calcified HC (arrows).

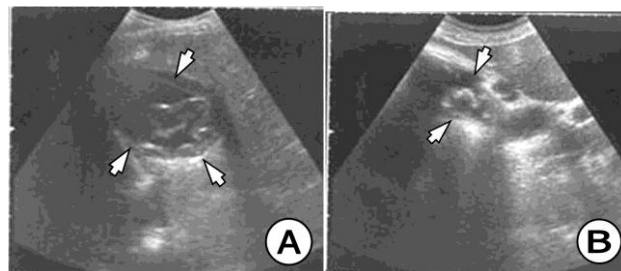


Figure11. (A and B) the US of the liver: A) The US clearly visualized inflamed HC of the liver with daughter cysts and debris (arrows). B) The US clearly visualized chronic inflammation of the calculus gallbladder, with hydatid daughter cysts into the lumen (arrows) as a result of the penetration of the liver HC into the biliary stem with content emptying.



Figure 12. (A and B) CT scan of the upper abdomen - axial view: A) In the right lobe of the liver segment VIII we can see, a multilocular inflamed HC with daughter cyst (arrowhead) with clearly demonstrates fistula between the HC and the biliary ducts (black arrows). In the segment VII, we can see a well-defined, smooth, thin-walled round solitary HC (white arrow). The common hepatic duct is widening with clear daughter into the lumen. B) In the gallbladder, we can see calcified HC (white arrows). Inflamed gallbladder with stones, debris and daughter cyst into the lumen.

In a female patient at the age of 27, with the US examination of the breast, an HC was diagnosed with typical sonographic features, round multilocular cyst with multiple daughter cysts with a diameter of 45mm.

In all patients, surgery was performed and postoperative verification of the diagnosis set by US and CT was obtained.

4. DISCUSSION

HD is endemic in the Middle East, Mediterranean countries, South America, North Africa, and Australia. (13) HD is an anthrozoosis caused by *Echinococcus* species. The *Echinococcus granulosus* is the most common response for cystic formation, follow by *Echinococcus multilocularis* (4). Humans represent intermediate hosts in parasite life cycle when occasionally ingest larvae. (1, 2) The ingested larvae settle in the liver and other organs through the blood and lymph. The host reacts to the cyst by forming a fibrous capsule (pericyst), which contains blood vessels that provide nutrients for the parasite. From the germinal layer, scolices, brood capsules, and daughter cysts are formed by endoproliferation. (1, 2)

Although HC disease can be found in almost all organs and tissues of the human body, it is most frequently seen in the liver (50–77%), the lungs (15–47%), the peritoneal cavity (8–12%), spleen (2–3%), the kidneys (1–4%), the brain (2%), the bones (1.5–2%), genitals (1%), muscles (0.5–1%) the spleen (0.5–8%), the kidneys (2–4%), and brain (up to 1%). (4, 5, 6, 14, 15)

Peritoneal hydatidosis is an uncommon finding. Only a few cases of primary peritoneal hydatidosis have been reported in the literature. (16, 17) Secondary peritoneal hydatidosis is more common than primary. (18) Peritoneal HC, either primary or secondary, represents an

uncommon but significant manifestation of this disease. (19) It is always secondary to traumatic or surgical rupture of a hepatic, spleen or mesenteric cyst. (18,20) CT is the modality of choice for these patients because it permits imaging of the entire abdomen and pelvis. The lesions are generally multiple and any type of HC can arise anywhere in the peritoneal cavity. (18, 21)

Clinically, nearly 30% of the patients with spleen HC are asymptomatic. Splenomegaly is the most frequent finding, which is incidentally determined. (9) The clinical symptoms caused by spleen HC mostly depend on the pressure effect of the cyst on the neighboring organs and the replacement of the neighboring organs. (22)

US and CT scans, alone or in combination, can establish a definite diagnosis of spleen HC in almost all cases. The US is the primary preferred diagnostic method because it is inexpensive, easy, and has a high diagnostic value. It is diagnostic because it shows the cystic structure of the lesion, the presence of daughter vesicles, and hydatid sand. (11) CT is usually the next step after a US diagnosis has been made. The main purpose is to visualize the relationship between the HC and the surrounding tissue. (11) Although CT scan is more sensitive than the abdominal US, non-calcified benign cysts without daughter cysts cannot be differentiated per se from other benign cysts either by CT or by the US. (11, 22, 23, 24) Differential diagnostic include epidermoid cyst, pseudocyst, solitary abscess, hematoma and cystic neoplasm of the lien. The diagnosis is easy when the HD has a multifocal localization and involves multiple organs and when daughter cysts are present. (21, 23, 24)

Although hydatid cyst disease can be present in all parts of the body, isolated renal involvement is a rare condition, comprising only 2–4% of all

cases. (25) The kidney is the most common location in the urogenital system. Renal HC causes symptoms with vague pain or a palpable mass in the lumbar region. (26, 27) In our case, the HC was identified incidentally. In our case, the lesion was located in the right kidney. Imaging methods are important for early diagnosis. Although the US is the most useful radiologic method, unenhanced CT has a high sensitivity for lesions. (12, 25, 26, 28, 29) The cyst can be unilocular or multilocular septal in case of active cysts. Renal hydatid cysts usually remain symptomless for years. (26, 29)

Isolated renal hydatid cyst disease may reside symptomless for a long time. Clinical presentations include fever, malaise, flank pain, palpable mass, hematuria, and hydaturia. (30,31) Although hydaturia which is characterized by the presence in urine of grape skin-like fragments, is the only pathognomonic sign that testifies of a ruptured hydatid cyst in the urinary tract, it has been seen only in 5%-25% of renal hydatid cyst disease. (30, 31) In our case, the patient had dull pain and palpable mass in the right flank with loss of weight and urinary symptoms such as nocturia and incontinence. However, there was no evidence of hydaturia. In the world literature, HD of the bladder is extremely rare, and in our practice so far we have not registered such a case.

Intracranial HD is rare, with a reported incidence of 1-2% of all cases with HD. (32) Cerebral HD is more common in the pediatric population. (33) Intracranial hydatid can occur anywhere within the brain but are more frequently located in the supratentorial compartment, especially in the middle cerebral artery territory. (32) The parietal lobe is the most common site with 69%, the other less common sites are a skull, cavernous sinus, eyeball, pons, extradural, cerebellum, and ventricles. (32, 34, 35).

The cerebral HC are benign and slowly growing cysts and present late when they increase in size and become large. A headache and vomiting were the most commonly reported symptoms in other series as well as in our patients. Other symptoms such as hemiparesis, seizures, visual field alteration and gait disorders, may vary with the location of the cyst. (35) Cerebral HC is commonly solitary. Multiple cerebral HC are rare. (34, 36)

Intracranial HC may also be classified as primary or secondary. In primary multiple cysts, each cyst has a separate pericyst with brood

capsule scolices and these originate from multiple larvae affecting brain after crossing the gastrointestinal tract, liver, lungs and right side of the heart without affecting them. (34) The secondary multiple cysts result from spontaneous, traumatic or surgical rupture of the primary intracranial HC and they lack brood capsule and scolices. (34) CT scans characteristically show HC as a spherical, well defined, an enhancing cystic lesion without peripheral edema. (10) The fluid density is generally equal to that of CSF. A fine rim of peripheral enhancement with perilesional edema may be seen in the presence of active inflammation. (10) In contrast to hydatid cysts, arachnoid cysts are not spherical in shape and not surrounded entirely by brain substance. Arachnoid cysts are extra-axial masses that may deform the adjacent brain. Cystic tumors of the brain could be differentiated by the enhancement of the mural nodule. When a pyogenic abscess shows a cyst like central necrotic area, peripheral edema is almost always present and the rim enhances intensely following contrast administration. (10, 32) The treatment of hydatid cyst is surgical and the aim of surgery is to excise the cyst in toto without rupture to prevent recurrence and anaphylactic reaction.

In the differential diagnosis of HC of the brain arachnoid cyst, porencephalic cyst, cystic tumor and abscess should be excluded. The arachnoid cyst is not round and is not surrounded by brain tissue like HC. The porencephalic cyst usually interacts with the ventricular system. The cystic tumor usually has a soft-tissue component, which is enhancement after i.v. application of contrast medium. The abscess, after i.v. administration of contrast medium shows the walls enhancement and has a surrounding edema. (32)

The involvement of the female genital tract in hydatid disease is extremely rare but the most important factor in its diagnosis is the awareness of the possibility of hydatid disease especially in the endemic area. (37,38) Ovarian involvement is also very rare and generally secondary to the peritoneal spread of daughter cysts due to rupture of a liver HC. (38,39) Ovarian HC is usually asymptomatic and they can be discovered incidentally or they may cause irritation or compression symptoms. (37,38,39) The ovarian lesion may be unilocular or contain daughter cysts that can give rise to a multiloculated appearance, and ovarian lesion should be considered in the differential

diagnosis of cystic pelvic masses such as cystadenoma or cystadenocarcinoma (37,38,39).

Radiological diagnosis of osseous HD is also difficult because of non-specific findings. Radiography usually reveals single or multiple osteolytic lesions and in some cases shows cortical thinning, osteosclerosis, and pathological fractures. (40, 41) Intraosseous foci of HD predominate in spongy bone tissues and consist of small, separate, and thin-walled cysts. While these cysts expand, they destroy the surrounding trabeculae and can reach a considerable size. (40, 42, 43) HC can mimic an image similar to an abscess or tumor, which is difficult to distinguish from a malignant bone tumor. Signs that mean much for a differential diagnosis include the osteoporosis zone, bone thickening of the affected bone, and calcification in the lesion. The association of the bone lesion with calcification in the surrounding soft tissue allows a reliable diagnosis of the HC. (40, 42, 43, 44, 45)

Ultrasound and CT scan of abdomen and pelvis may help to determine the exact location and nature of a cystic pelvic mass, its relationship with adjoining structures, vascular invasion, operability and also exclude hydatid disease elsewhere. (44, 45)

Spinal HC can be manifested by different clinical features. It is considered to be relatively silent, slowly progressive disease with a latent period of many years. (40, 46) Early symptoms appear when the small cyst expands and produces significant mechanical compression on surrounding vital structures. At this stage, intermittent pain can be present as a symptom. As the disease progresses and the cyst enlarges, the patient starts suffering from a persistent backache, gradually leading to the severe neurologic deficit and varying degrees of weakness of limbs. (40, 46, 47, 48) When HD affects the spine, tuberculous spondylitis appears as the most common differential diagnostic problem. The absence of destruction on the discal surfaces of the vertebrae and the spread of the disease through subperiosteal and subligamentous pathways are typical of vertebral HD. (46,47)

Muscle HD is rare, possibly because of muscle lactic acid content and muscle contraction, two factors that likely prevent cyst growth in striated muscle. Clinically, the early stage of muscular HD is asymptomatic. The symptoms and signs of HC disease depend on the involved organ,

site of localization, effect on the adjacent tissue, complications after rupture. A palpable mass is the most constant clinical finding of HD affecting soft tissues, and the clinical manifestations are caused by compression of the organ involved. (49, 50) Muscle HD most often manifests as a slow-growing soft-tissue tumor and can imitate myositis or a calcified hematoma.

A common complication of the hydatid cyst of the liver is its penetration into the biliary ducts. The US, and especially CT study gives enough information for successful diagnosis and further successful surgery. (51) CT examination enables excellent space presentation of pathologic process, which is especially important for an operator for surgery planning approach, techniques, and procedures which will be applied for improving the pathological process. Primary hydatid cyst of the gallbladder is an extremely rare entity. (52, 53, 54) There are reports of the gallbladder daughter cysts secondary to liver cysts. (55, 56)

Gold standard treatment is the total excision of the cyst, avoiding its rupture and spillage; anaphylactic shock is a major risk connected. Surgical treatments include complete cyst excision in most of the patients. Nephrectomy, cholecystectomy, ovarian cystectomy, distal pancreatectomy, and splenectomy, were performed whenever cysts invaded these organs.

Indications for surgery include large cysts with multiple daughter cysts, superficial location amenable to rupture, cysts exerting pressure on adjacent organs, and cysts in ectopic locations such as seminal vesicle, brain, bones, spleen, kidneys.

5. CONCLUSION

Echinococcosis can appear at any site of the human body, therefore in endemic areas, whenever we have cystic formations or unidentified tumor masses in the differential diagnosis, HD should also be considered.

US imaging has great value in the screening of HD, it is cheap, easily accessible, and simple to perform a method, with high diagnostic accuracy. CT provides precise topographic and morphological features of the pathological substrate, with high sensitivity, specificity, and accuracy.

We can conclude that imaging methods have a great diagnostic value in HD with atypical

localization, in its verification and preoperative evaluation.

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